

Incidence of Palatal Fistula after One-Stage Palatoplasty and Factors Influencing the Fistula Occurrence

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Background: Cleft palate repair may be compromised by a number of complications, most commonly the development of a fistula. Fistulas are related to an increased rate of hypernasal speech, articulation problems, and food or liquid regurgitation from the nose. Fistulas also tend to recur after a secondary repair to address the fistulas. This study reviews the rate of fistula in our craniofacial center after a one-stage cleft palate repair; and to determine whether, cleft type, age at repair, type of cleft repair, hemoglobin level presurgery, and patients nutritional state influence the risk of fistula occurrence.

Patient and Method: A retrospective analysis was performed on medical records of 93 patients who underwent palate repair between January 2012 to October 2013. All consecutive cleft (lip and) palate patients are included. Bivariate analysis was performed to identify the predictors of fistula formation.

Result: Ninety-three patients (50 male and 43 female) underwent one-stage palatoplasty. Cleft palate fistulas occurred in 19 of 93 patients (20,4%). The age of the patients at the time of repair ranged from 9 to 144 months (mode 18 months). All palate repairs were done in one stage, using either the two flap (N=66), Wardill-Kilner (N=24), Furlow (N=2), and Langenback (N=1) techniques. No significant influence was found related between age at the time of repair (p 0.789), body weight (p 0.725), Hemoglobin value (p 0.295), and type of cleft (p 0.249) to the rate of fistula occurrence.

Summary: This study found no association between , body weight, preoperative hemoglobin value, and the type of cleft to the rate of fistula following cleft palate surgery.

Keywords: *cleft palate, palatal fistula, palatoplasty, factors affecting fistula*

Latar Belakang: Hemangioma merupakan tumor yang sering ditemukan pada bayi, bersifat tumbuh cepat setelah kelahiran dan regresi perlahan di masa anak-anak. Sumbing bibir merupakan kelainan yang umum ditemukan di Indonesia. Kedua kelainan ini banyak didapatkan di populasi Asia. akan tetapi, kejadian simultan kedua anomali ini jarang ditemukan.

Pasien dan Metode: Sebuah kasus sumbing bibir bilateral dgn hemangioma pada sisi kiri bibir dan kulit sekitarnya sampai ke mukosa dan prolabium yang telah menjalani operasi cheiloplasty di usia 9 bulan. Desain cheiloplasty dibuat melewati area hemangioma dan dengan hemostasis yang baik untuk mencegah pendarahan masif.

Hasil: Hemangioma ditinggal sebagian saat operasi cheiloplasty. Tidak ada pendarahan saat dan setelah operasi. Dua tahun pasca operasi, ditemukan garis parut yang halus dengan bentuk bibir yang baik dan tidak ada pertumbuhan hemangioma.

Ringkasan: Hemangioma dan sumbing bibir jarang ditemukan pada lokasi yang sama. Waktu operasi yang tepat sangat penting karena adanya potensi involusi hemangioma. Dengan mempertimbangkan aspek psikososial, operasi dilakukan di usia pasien 9 bulan. Dua tahun pasca operasi, tidak ditemukan pertumbuhan hemangioma atau deformitas tulang dan bibir. Jaringan parut tumbuh dengan estetik yang baik, tidak berbeda dengan pasien sumbing tanpa hemangioma..

Kata Kunci: *hemangioma, cleft lip, scar, deformity*

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Cleft lip and palate is one of the most occurring congenital anomalies worldwide affecting babies 1:700 live birth.¹ Cleft lip and palate together occurs every 1:1289 live births, cleft lip alone occurs 1:1000

live births, more commonly among the boys and three times more frequent than the cleft palate alone. Cleft palate alone occurs 1: 2500 live births with higher incidence among the female babies.² Cleft lip and palate is the most frequent diagnosis accounting for 46% of all the cleft populations.³

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Cleft palate repair aim to attain the development of normal speech without significantly impairing maxillary outgrowth, as well as minimizing hearing loss and middle ear complications.⁴ In managing patients with cleft palate, the most controversial issues include the timing of surgery, speech development, and facial growth.⁵ The ideal age for cleft palate surgery is usually 3 to 18 months. Speech and hearing are improved by cleft palate repair before 24-month of age. In Cipto Mangunkusumo Hospital, Jakarta, speech outcome give better results when cleft palate repair is performed before 2-year-old.⁶ Delayed closure (after five years) is associated with retardation of maxillofacial growth.⁷

The incidence of fistulas after palatoplasty ranges from 3 to 38%.⁸ Larger studies report rates in range of 10-20%.⁹ Palatal fistulas may present as asymptomatic holes or may cause such symptoms as speech problems, nasal regurgitation of fluids, or difficulty in maintaining oral hygiene. The most common locations for fistulas are at the region around the incisive foramen, at the posterior nasal spine, and the uvula.

All postoperative fistulas are found to be contributed to either failure of healing or breakdown of the original cleft palate repair. The incidence is highly variable although the primary cause remains the same in most, which is due to closure under tension and infection.¹⁰ Failure of healing of the palatal wound post repair may lead to scarring and fistula.¹¹ Anatomically, the size of cleft size as well as the technique of repair are factors which influence fistula occurrence.¹² Cleft size affect the difficulty of surgical repair and, thus, indirectly affect postoperative maxillary growth.¹³ Facial and palatal growth retardation following cleft repairs is said to be due to destruction of blood supply and scar formation.¹⁴

In Indonesia, only a few studies have investigated the incidence of fistulas post palate repair. Our craniofacial team has implemented the one-stage repair for cleft palate repair for many years, however, the final outcome of the protocol was never properly reviewed. This study aims to detect the incidence of palatal fistula post palatal repair in our institution. Another study was conducted by our division in conjunction with Adam Malik Hospital in Medan

which found that dental caries is one of the the predisposing factors for palatal fistula formation.¹⁵ Using data from the medical records, the incidence of palatal fistula on patients post palate repair due to cleft palate is tabulated, and factors of interests analyzed to see if those factors are contributable to the formation of fistulas.

PATIENT AND METHOD

The clinical records of patients who underwent primary cleft palate repair between January 2012 to October 2013 were retrospectively reviewed. Patients and craniofacial team are from the Ciptomangunkusumo Hospital Jakarta, Indonesia. Patients included are those who have a complete operative and follow-up medical records with a minimum follow-up period of at least 2 months after the time of primary palate repair.

Other variables of interest are noted for each patient, which include gender, age at the time of primary repair, type of cleft, body weight, and hemoglobin level presurgery. The surgical technique used is also noted, with the primary outcome of the study as whether or not patients developed a palatal fistula. Age of the patients at the time of surgery is divided into two groups, younger than 2 years old and older than 2 years old.¹⁶ Body weight is classified based on WHO-CDC growth chart: normal weight or underweight, at the time of surgery.¹⁷ Hemoglobin level is categorized into either normal or anemic using the cutoff level of anemia for pediatric patients.¹⁸

Cleft of the palate is defined as either complete, partial, or isolated. Based on the Veau classification, the type of cleft that is included in the study are those of class III and IV, which is the complete and incomplete or isolated cleft lip and palate. The surgical techniques used for palate repair follows the protocol of the craniofacial team in this institution. Bardach two-flap palatoplasty is selected for cases of complete cleft palate, the V-Y pushback palatoplasty is used in cases of partial or isolated cleft palate, whilst other techniques such as the Furlow and Langenbeck repair are occasionally used in selected cases.

All patients with either complete cleft of the lip and palate, partial cleft palate, or isolated cleft palate who underwent the one-stage palate repair in our unit, during the study time, were included. The age at the time of primary repair is limited up to the adolescent age. Adult patients, patients with missing or incomplete medical records, and patients with no follow up are excluded from the study.

Initial diagnosis was established on the basis of history and clinical examination of the oral cavity. Postoperatively, patients were given prophylactic antibiotics and analgesics in the form of suspension or intravenous. Parents were educated on how to administer food and what to avoid postoperatively, such as liquid diet was to be given using a spoon, finger (or other object) suckling into the mouth is prohibited. Diagnosis of palatal fistula is obtained from physical intraoral examination post surgery.

Only symptomatic fistulas of the hard palate which require revisional surgery are included. All relevant data was collected and analyzed using SPSS version 20.0. Statistical analysis is done using the Chi-square test and Fischer exact test . A *p* value less than 0.05 is considered statistically significant in all instances

RESULT

There is a total of 134 patients who underwent palatoplasty in between January 2012 until October 2013 in Ciptomangunkusumo Hospital, Jakarta. 38 of those have incomplete data records, or failed to show up for a follow-up. These patient were not included in the investigation, leaving 93 patient records for analysis. Syndromic patients are also not discussed in this study.

Table 1. Characteristics of study populations and incidence of fistula

	All		Fistule			
	N = 93	%	Yes		No	
	N	%	N	%	N	%
Sex						
Male	50	53,8%	8	16%	42	84%
Female	43	46,2%	11	25,6%	32	74,4%
Age						
> 2 y.o	27	29%	6	22,2%	21	77,8%
< 2 y.o	66	71%	13	19,7%	53	80,3%
Body weight						
Underweight	13	14%	3	23,1%	10	76,9%
Normal	80	86%	16	20%	64	80%
Hemoglobin						
Anemia	13	14,0%	1	7,7%	12	92,3%
Normal Hb	80	86%	18	22,5%	62	77,5%
Type of cleft						
Complete cleft lip and palate	64	68,8%	11	17,2%	53	82,8%
Incomplete cleft lip and palate	29	31,2%	8	27,6%	21	72,4%
Technique of palatoplasty						
Two flap	66	71%	12	18,2%	54	81,8%
V-Y pushback	24	25,8%	4	16,7%	20	83,3%
Furlow	2	2,2%	2	100%	0	0%
Langenbeck	1	1,1%	1	100%	0	0%

Out of the 93 analyzed patients, 19 out of 93 patients (20,4%) had palatal fistula. The size of the fistula was not noted in the records. The baseline characteristics of the study population is summarized in Table 1. These patients included 50 boys (53,8%) and 43 girls (46,4%) whose median age was 18-month-old (range 9 to 144 month-old) at the time of repair. Body weight is categorized according to WHO growth chart¹⁷, defined as either normal or underweight, based on the z-score interpretation. Eighty patients had normal weight (86%) while 13 patients were underweight (14%). Anemia is defined as hemoglobin (Hb) level of lower than 11,1-11,9 g/dL.¹⁸ Anemia was found in 13 out of 93 patients (14%). Complete cleft lip and palate was true in 64 cases (68,8%), while in the remaining 29 cases (31,2%) had incomplete cleft lip and palate.

Four techniques were used to repair the cleft palate with the majority of patients underwent the Bardach's two flap palatoplasty (66 patients or 71%), followed by Wardill-Kilner V-Y pushback in 24 patients (25,8%), Furlow palatoplasty in 2 patients (2,2%), and Langenbeck in 1 patient (1,1%).

Fistula rates were grouped by gender, age, body weight, hemoglobin value, and type of cleft (Table 2). The variables are categorized into two categorical data, analyzed using the Chi square bivariate analysis for each variable. The rate of palatal fistula is higher among the female patients (25,6%) than in male patients (16%), however there was no statistically significant differences in sex distribution (p 0.253). No significant difference were found on the risk of fistulas when tested in regard to the age at the time of repair (p 0.789), body weight (p 0.725), value of hemoglobin p 0.295), and the type of cleft (p 0.249).

Table 2. Bivariate Analysis of Gender, Age, Body Weight, Hemoglobin value, and Cleft type

	N = 19	Patient developing fistula	P value	Risk Ratio	CI 95%
Sex					
Male (n = 50)	8	16%	0.253	0.625	0.277 - 1.412
Female (n = 43)	11	25,6%			
Age					
> 2 y.o (n = 27)	6	22.2%	0.789	1.128	0.479 - 2.659
< 2 y.o	13	19.7%			
Body weight					
Underweight (n = 13)	3	23.1%	0.725*	0.623	0.280 - 1.384
Normal (n = 80)	18	20%			
Hemoglobin					
Anemia (n = 13)	1	7.7	0.295	1.154	0.390 - 3.414
Normal Hb	80	22.5%			
Type of cleft					
Complete CLP (n = 64)	11	17.2%	0.249*	0.342	0.050 - 2.347
Incomplete CLP (n = 29)	8	27.6%			

CLP = cleft lip and palate

*Using Fischer exact test

DISCUSSION

The incidence of palatal fistula post palatoplasty have been described in many previous studies. This study reviews the overall fistula incidence in our institution by including all palate repair patients during a 21-month period, the rate of fistula is found to be 20,4%. From the literature, fistulas post palatal repair have been associated to gender, type of cleft, technique of repair, the cleft size, surgeon's experience, and age at the time of repair. The variables were investigated in this study.

This study detected a relatively high rate of fistula following palate repair. This may due to the relatively small number of cases and multifactorial etiologies. From the data available in this study we evaluated the effect of gender, age at the time of repair, body weight, hemoglobin value, and the type of cleft; on how they affect risk of fistulas postrepair.

Previous studies suggests that the risk of fistula formation is higher as width of cleft increases.⁵ Cleft gap affects the difficulty of surgical repair, thus indirectly influence scar formation and postoperative maxillary growth.⁸ Lower rate of clinically significant fistula is attributed to early soft palate repair, with smaller secondary clefts allowing repair with minimal dissection and disruption of vascularity.¹⁹ It has also been reported that fistula rate is higher among patients with wider clefts, regardless of the type of surgical repair²⁰.

Regarding the technique of repair, the majority of patients in this study underwent one of the three techniques of one-stage repair, the two-flap palatoplasty for complete cleft lip and palate, and the Veau-Wardill-Kilner or V-Y pushback palatoplasty for incomplete cleft lip and palate. The Furlow and Langenbeck techniques were also performed, in 2 and 1 patient respectively. Analysis for the different techniques used was not conducted in this study. Previous studies stated that the choice of the technique of repair is associated to the rate

of fistula.²¹ The two-flap palatoplasty was reported to have a lower rate of fistula because it effectively provides tension-free, multilayer repairs.²² The V-Y pushback that was used in cases of incomplete cleft lip and palate has the advantage of lengthening the palate and repositioning the levator muscle in a more favorable position,²³ and the disadvantage is a higher rate of fistula in repair of complete cleft palate than the other techniques because it provides only a single nasal mucosa layer anteriorly.²⁴ The Furlow opposing Z-plasty, is effective for the primary closure of a submucous cleft palate and the secondary correction of marginal velopharyngeal insufficiency.²⁵⁻²⁶ The Langenbeck technique is the oldest technique which is still widely used, commonly for an incomplete cleft of the secondary palate without the presence of a cleft lip and alveolus.²³

In this study the occurrence of fistula following cleft palate repair is found to be irrelevant to the age at the time of repair, nutritional status based on body weight, hemoglobin value, and the type of cleft. Several prior studies have reported that there is no association between fistula occurrence and gender, age at the time of palatoplasty, and the operating surgeon; however these were done among small population size.^{24,27}

Determining the ideal age for time of repair is mainly based on the expectation of good speech outcome, prevention of velopharyngeal insufficiency, maxillofacial growth disturbance, and hearing disorder.²⁸ Recent study showed speech were equally good when the repair is performed between 12-18 months of age.⁴ Another study shows that earlier age of repair (younger than 7-month-old) does not result in improved speech outcome over palatoplasty performed later in infancy.²⁹ Previous study suggest that there were statistically greater overall and persistent fistula rates in the late closure palatoplasty, even it does not explain the exact etiology.⁴

The type of cleft has been reported to have association with the type and location of the fistula. The higher incidence of anterior fistula was found in patients with a bilateral cleft lip and palate compared with those with a unilateral cleft lip and palate or incomplete cleft of the hard palate.⁸ In this study, the location of fistula was not well recorded. The size and location of palatal fistulas are functionally important because compensatory articulatory deficits develop that are difficult to correct if not closed early during speech formation.²⁸

CONCLUSION

In this study, there were no association between fistula rates according to gender, body weight, hemoglobin value, type of cleft. However, the risk of palatal fistula always persists even in the best technique or in hands of the best surgeons. It is our responsibility to keep evaluating results and find alternatives to obtain the optimum management for our patients. This study is still lacking number of cases and data to obtain sufficient data the records have to be properly written and should consist of information that could be used for further investigation. Prospective study in larger population should be considered to determined the exact etiology of palatal fistula.

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REFERENCES

1. Olasoji HO, Ukiri OE, Yahaya A. Incidence and etiology of oral clefts, a review. *Afr J Med.* 2005;34:1-7.
2. Millard DR. *Cleft craft: the evolutions of its surgery-volume I: The unilateral deformity.* Baltimore: The Williams and Wilkins Co; 2007.
3. Thorne CH, Beasley RW, Aston SJ, et al. *Grabb and Smith's plastic surgery,* ed 6. Philadelphia: Lippincott Williams & Wilkins; 2007.
4. Rohrich RJ, and Rowsell AR. Timing of hard palatal closure: a critical long-term analysis. *Plast Reconstr Surg.* 1996;98(2):236-46.
5. Shah SA, Khan F, Bilal M. Frequency of fistula formation after two stage repair of cleft palate. *JKCD.* 2011;2(1).
6. Djoenaedi I, Handayani I, Wahyuni LK, Bangun K. Speech outcome evaluation after two-flap palatoplasty in plastic surgery division Ciptomangunkusumo Hospital: a retrospective study. *Jurnal Plastik Rekonstruksi.* 2012;1(4):409-416.
7. Graber TM. A cephalometric analysis of the developmental pattern and food morphology in cleft palate. *Angle Orthod.* 1949;19:91-102.
8. Phua YS, and de Chalain T. Incidence of Oronasal Fistulae and Velopharyngeal Insufficiency After Cleft Palate Repair: An Audit of 211 Children Born Between 1990 and 2004. *The Cleft Palate-Craniofacial Journal.* 2008;45(2):172-178.
9. Becker M, and Hansson E. Low rate of fistula formation after Sommerlad palatoplasty with or without lateral incisions: An analysis of risk factors for formation of fistulas after palatoplasty. *Journal of Plastic, Reconstructive & Aesthetic Surgery.* 2013;66(5):697-703.
10. Sadhu P. Oronasal fistula in cleft palate surgery. *Indian J Plast Surg.* Oct 2009;42(Suppl):S123-S128.
11. Musgrave_RH, and Bremner JC. Complications of Cleft Palate Surgery. *Plastic & Reconstructive Surgery & the Transplantation Bulletin.* 1960;26(2): 180.
12. Tahir M, Tahmeedullah, Khan AT, Rubina. Speech result of Furlows soft palate repair. *JPMI.* 2004;4:607-13.
13. Liao YF, Prasad NKK, Chiu YT, et al. Cleft size at the time of palate repair in complete unilateral cleft lip and palate as an indicator of maxillary growth. *Int J Oral Maxillofac Surg.* 2010;39:956-61.
14. Cohen M, Smith Bonnie E. Cleft lip and palate In: Goldwyn Robert M, Cohen M. *The unfavorable result in Plastic Surgery Avoidance and treatment* 3rd ed. Philadelphia: Lippincott Williams and Wilkins 2001; 303-33.
15. Utama D, Buchari F, Sudjatmiko G. The incidence of palatal fistula postpalatoplasty in children with dental caries: A multi centre study. *Jurnal Plastik Rekonstruksi.* 2013;2(2):78-83.
16. Dorf D, and Curtin, JW. Early cleft palate repair and speech outcome. *Plast Reconstr Surg.* 1982;70:75.
17. 2000 CDC Growth Charts for the United States: Methods and Development . Vital and health statistics. Series 11, Data from the National Health Survey. 2002;246.
18. Behrman, Richard E. *Nelson Textbook of Pediatrics : 17th Edition.* Philadelphia: WB Saunders; 2000:1462.
19. Muzaffar AR, Byrd HS, Rohrich RJ, Johns DF, LeBlanc D, Beran SJ, et al. Incidence of cleft palatal fistula. An institutional experience with two stage

20. Mak ASY, Wong WH, Or C, Poon AM. Single surgeon's experience with Furlow palatoplasty and the fistula rate. *Surgical Practice*. 2006;10:138-42.
21. Amaratunga NA. Occurrence of oronasal fistulas in operated cleft palate patients. *J Oral Maxillofac Surg*. 1988;46:834-8.
22. Wilhelmi BJ, Appelt EA, Hill L et al. Palatal fistulas: rare with the two-flap palatoplasty repair. *Plast Reconstr Surg*. 2001;107(2).
23. Leow AM, Lo LJ, Palatoplasty: evolution and controversies. *Chang Gung Med J*. 2008;31(4). 335-45.
24. Cohen SR, Kalinowski J, LaRossa D, Randall P. Cleft palate fistulas: a multivariate statistical analysis of prevalence, etiology and surgical management. *Plast Reconstr Surg*. 1991;87:1041-7.
25. Chen PK, Wu J, Hung KF, Chen YR, Noordhoff MS. Surgical correction of submucous cleft palate with Furlow palatoplasty. *Plast Reconstr Surg*. 1996;97:1136-46.
26. Chen PK, Wu JT, Chen YR, Noordhoff MS. Correction of secondary velopharyngeal insufficiency in cleft palate patients with the Furlow palatoplasty. *Plast Reconstr Surg*. 1994;94:933-41.
27. Emory RE, Clay RP, Bite U, Jackson IT. Fistula formation and repair after palatal closure: an institutional perspective. *Plast Reconstr Surg*. 1997;99:1535-8.
28. Rohrich R J., Love E J., Byrd HS, and Johns DF. Optimal timing of cleft palate closure. *Plast Reconstr Surg*. 2000;106(2):413-422.
29. Kirschner RE, Randall PMD, Cleft Palate Repair at 3 to 7 Months of Age. *Plast Reconstr Surg*. 2000;105(6):2127-2132.